

Vocal cord dysfunction after pediatric cardiac surgery: A prospective implementation study



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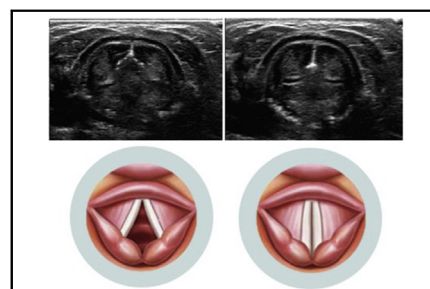
ABSTRACT

Objective: To determine the incidence, outcomes, and evaluate diagnostic modalities for postoperative vocal cord dysfunction (VCD) following cardiothoracic surgery in children.

Methods: A prospective mixed-methods study using principles of implementation science was completed. All patients undergoing surgery involving the aortic arch, ductus, or ligamentum arteriosum and vascular rings from September 2019 to December 2020 were enrolled. Patients underwent speech pathology assessment, laryngeal ultrasound, and flexible direct laryngoscopy.

Results: Ninety-five patients were eligible for inclusion. The incidence of VCD ranged from 18% to 56% and varied according to procedure group. VCD occurred in 42% of neonates. Repair of hypoplastic aortic arch was associated with increased risk of VCD (57%; $P = .002$). There was no significant difference in duration of intubation, pediatric intensive care unit stay, or hospital stay. Forty percent children were able to achieve full oral feeding. Children with VCD were more likely to require nasogastric supplementary feeding at discharge (60% vs 36%; $P = .044$). Sixty-eight percent of patients demonstrated complete resolution of VCD at a median of 97 days postoperatively. Laryngeal ultrasound and speech pathology assessment combined had a sensitivity of 91% in comparison to flexible direct laryngoscopy.

Conclusions: VCD occurred in one-third and resolved in two-thirds of patients at a median of 3 months following cardiac surgery. Aortic arch repair carried the highest risk of VCD. VCD adversely influenced feeding. Forty percent of patients achieved full oral feeding before discharge. VCD did not delay intensive care unit or hospital discharge. Speech pathology assessment and laryngeal ultrasound combined was reliable for diagnosis in most patients and was more patient friendly than flexible direct laryngoscopy. (JTCVS Open 2022;11:398-411)



Laryngeal ultrasound reliably demonstrates abduction and adduction of the vocal cords.

CENTRAL MESSAGE

VCD occurs in one-third and resolves in two-thirds of patients following cardiac surgery. Ultrasound is reliable for diagnosis in most patients.

PERSPECTIVE

VCD occurs in one-third and resolves in two-thirds patients at a median of 3 months following cardiac surgery. VCD adversely influences feeding. VCD does not delay ICU or hospital discharge. Speech pathology assessment and ultrasound combined is reliable for diagnosis in most patients.

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Abbreviations and Acronyms

ENT	= ear, nose, throat
FDL	= flexible direct laryngoscopy
LUS	= laryngeal ultrasound
NGT	= nasogastric tube
PICU	= pediatric intensive care unit
RLN	= recurrent laryngeal nerve
VCD	= vocal cord dysfunction
VCP	= vocal cord palsy

 Video clip is available online.

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Children undergoing surgery involving the aortic arch, pulmonary arteries, and ductus or ligamentum arteriosum are at risk of vocal cord dysfunction (VCD) due to the vulnerability of the recurrent laryngeal nerve (RLN). The incidence of VCD ranges widely from 4.5% to 59%.¹⁻³ This wide variation may reflect a reliance on retrospective data but could also reflect the lack of a standardized approach to screening tempered by the invasive nature of endoscopic evaluation. The clinical course and resolution of VCD remain poorly understood.

The consequences of VCD include prolonged tube feeding, aspiration-induced lung injury, voice abnormalities, increased hospitalization and mechanical ventilation times, increased frequency of respiratory infections, bronchopulmonary dysplasia, reactive airway disease, and airway intervention.⁴⁻⁸ This has resulted in some units routinely performing invasive assessment of all cases at risk of RLN injury.^{2,9}

The gold standard diagnosis of VCD relies on endoscopic laryngeal evaluation, which can be associated with pain, discomfort, and requires patient cooperation.¹⁰ Transcutaneous laryngeal ultrasonography (LUS) is a relatively novel but promising means of diagnosing VCD noninvasively.^{2,11} It is reproducible, safe, and well tolerated by patients and families.^{2,12,13}

We aimed to evaluate the incidence and outcomes of VCD for children undergoing cardiothoracic surgery for congenital heart disease and to evaluate the sensitivity and specificity of LUS as a diagnostic modality compared with the gold standard flexible direct laryngoscopy (FDL). Principles of implementation science were utilized to refine our existing protocol to guide assessment and early feeding strategies.

METHODS

The study was approved by the Hospital Research Ethics Committee (HREC/18/QCHQ/49022). Written informed consent was obtained from the parents before enrolling participants in the study. The study was registered on the Australia and New Zealand Clinical Trials Registry (ACTRN12620000835943).

Participants

Patients aged 0 day to 18 years undergoing at-risk surgery from September 2019 to December 2020 were eligible to be enrolled on the study pathway. Eligibility required planned surgery on the aortic arch, ligamentum arteriosus, or ductus arteriosus. Children with preexisting VCD were excluded.

Study Design

This prospective, mixed-method implementation study was completed at a single tertiary pediatric hospital providing a statewide cardiac service. Detailed research methods and implementation protocol have been described previously.¹⁴ Our aims included simultaneous implementation of a robust and clear clinical pathway for at-risk children using the principles of implementation science and the Consolidated Framework for Implementation Research and guided by an implementation expert (G.M.).¹⁵⁻¹⁷ The clinical pathway (Figure 1) was designed and implemented by a team involving cardiac and ear, nose, throat (ENT) surgery, radiology, speech pathology, dietetics, cardiology, and nursing staff.

Statistical Analysis

Patient and operative variables were summarized, with continuous data expressed as median (interquartile range) and assessed for differences by VCD status. Differences in continuous and categorical variables were assessed with the Wilcoxon rank sum test and Fisher exact test, respectively. For statistical analysis, primary surgical procedures were stratified into 4 groups (Appendix E1 and Table E1). The sensitivity, specificity and other diagnostic measures were calculated for combined assessment of LUS plus speech pathology, as well as each modality alone, compared with the gold standard FDL.

Diagnostic Pathway

Assessment for VCD was delayed until discharge from the pediatric intensive care unit (PICU). Patients initially underwent evaluation by a speech pathologist and LUS in the high-dependency area of the ward. Irrespective of clinical signs, all patients were assumed to have VCD and were orally fed using specific feeding practices (Figure 1) until FDL assessment was performed. FDL was performed after transition to a lower intensity ward environment (Video 1). Because FDL was the final step in the diagnostic pathway, the speech pathologist, ultrasonographer, and radiologist were blinded to the FDL results. The consultant ENT surgeon who reviewed the video recordings of the FDL was blinded to the LUS results. Both paresis and palsy were categorized as VCD.

Influence of COVID-19 Pandemic

Because of the COVID-19 pandemic, hospital restrictions were placed on aerosol-generating procedures and patients could not undergo FDL for 3 months during the study period. Study participants underwent assessment by speech pathology and LUS only.

Assessment by a Speech Pathologist

Each assessment included screening of oral anatomy and clinical indicators of VCD, including qualitative assessment of cry (where age appropriate), vocalizations, stridor, and cough. Oropharyngeal feeding and swallowing skills were then assessed. Neonates and infants were either bottle or breastfed according to parental preference. Positional and postural

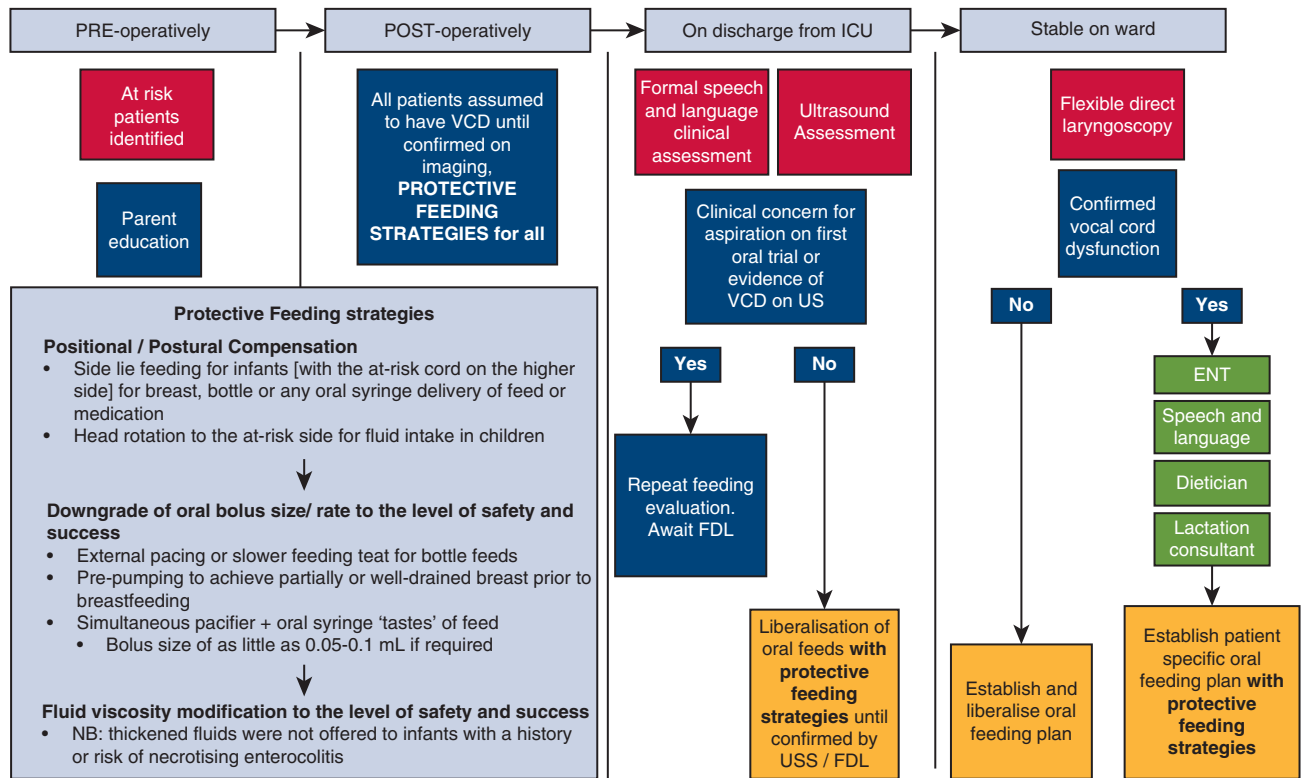
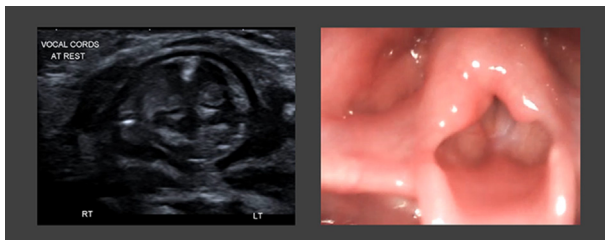


FIGURE 1. Clinical pathway for diagnosis and management of a child at risk of postoperative vocal cord dysfunction (VCD). ICU, Intensive care unit; US, ultrasound; FDL, flexible direct laryngoscopy; ENT, ear, nose, throat.

changes were utilized for fluid intake, including head-turn (toward the compromised side) to direct the bolus toward the opposing pharyngeal channel, and a side-lye position (with the at-risk cord on the higher side) for breast- or bottle-feeding infants. Patients were assessed for signs of aspiration, including eye widening or watering, coughing, or vocal or respiratory changes during or after feeding. Oxygen saturation, heart rate, temperature, and chest radiographs were also monitored. If there was no evidence of aspiration, feeding was gradually liberalized to a patient-specific oral feeding plan involving 1 or more of the following feeding modalities: syringe, oral, bottle, breastfeeding in combination with supplementary nasogastric tube (NGT) feeding as required. Daily review by a speech pathologist permitted gradual weaning from NG to partial or full oral feeding (including bottle- or breastfeeding) as tolerated.



VIDEO 1. Gareth Lloyd, consultant ear, nose, throat surgeon, discusses the diagnostic modalities utilized in the study, with demonstrations of normal and abnormal vocal cord function. Video available at: [https://www.jtcvs.org/article/S2666-2736\(22\)00246-7/fulltext](https://www.jtcvs.org/article/S2666-2736(22)00246-7/fulltext).

FDL

Beside FDL was performed by an ENT surgeon using a Storz 2.9 mm CMOS Video Rhino-Laryngoscope 11102 CM (Karl Storz SE & Co). Video images were reviewed by an ENT consultant. Administration of sedatives were avoided when possible.

Ultrasonography

LUS was performed by 3 pediatric sonographers using a Philips Epiq 7G platform (Philips Healthcare). Static images were recorded with the cords abducted (Figure 2, A) and adducted (Figure 2, B). Cine-loops were recorded for review by a single consultant radiologist.

VCD was diagnosed on LUS or FDL when there was asymmetric or incomplete adduction of one or both cords.

Follow-up

Patients with VCD had follow-up LUS at 3 months postdischarge and an ENT review with FDL at 6 and 12 months. Patients were also followed-up by a speech pathologist as required.

Outcome Measures

In addition to demographic and perioperative data, the following variables were collected: days to first oral feeding trial, days to liberalization to patient-specific feeding plan, days of NGT feeding, mode of feeding pre-operatively and on discharge, gastrostomy, confirmed or suspected aspiration events during admission, tracheostomy, and readmissions to hospital. The mode of feeding was categorized as fully oral feed, NGT supported (>50% of daily nutrition orally), NGT predominant (<50% of daily nutrition orally), and nil by mouth.

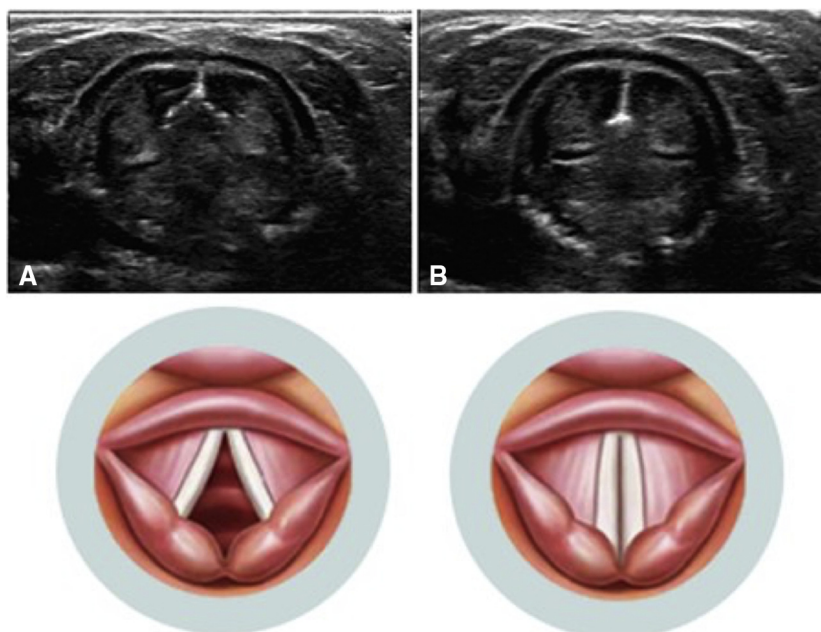


FIGURE 2. Static images demonstrating cords abducted (A) and cords adducted (B) on laryngeal ultrasound.

RESULTS

One hundred twelve patients were consented for the study and 13 (12%) were subsequently excluded; 8 did not meet the inclusion criteria and 5 died before assessment (see CONSORT diagram Figure E1). Four patients with complex multilevel airway disease were also excluded because these patients could not be assessed and managed using the study protocol: a neonate with Xq24 deletion

UBE2A deficiency who was tracheostomized for multilevel airway obstruction; a neonate with Pierre-Robin syndrome and 22q11 and multilevel airway disease; a neonate with Cri du Chat syndrome who did not make any vocal or feeding effort; and a 2-year old child with an undiagnosed genetic abnormality with preexisting laryngomalacia, microcephaly, cleft lip and palate, micrognathia, macroglossia, anterior anus, and hypertelorism who could not be fed orally. Baseline characteristics of patients are summarized in Table 1. Ages ranged from day 1 of life to 10 years. There were 51 (54%) neonates. Primary surgical procedures and intraoperative data are summarized in Table 2.

TABLE 1. Baseline characteristics (N = 95)

Characteristic	Result
Demographic	
Male	52 (55)
Median weight at surgery (kg)	3.6 (3.1-6)
Median age at surgery (d)	26 (10.5-126)
Age group	
Neonates	51 (54)
Infants	30 (32)
Children	14 (15)
Preoperative	
Ventricular physiology	
Biventricular	86 (91)
Univentricular	7 (7)
Indeterminate	2 (2)
Prematurity (<37 wk gestation) (%)	6/51 (12)
Chromosomal abnormalities	
Trisomy 21	4
22q11 deletion	1
Turner syndrome	1
Other chromosomal abnormalities	6

Values are presented as n (%) or median (interquartile range).

Incidence

Overall, VCD was present in 30 patients (32%). Left VCD occurred in 27 (90%) and right VCD in 3 (10%) patients (ie, vascular ring with right-sided aortic arch and left ligamentum [n = 2] and hypoplastic right aortic arch with division of left ligamentum arteriosum [n = 1]). The incidence of VCD ranged from 18% to 56% and varied according to procedure group with the highest incidence in patients undergoing repair of hypoplastic aortic arch via median sternotomy (Table 3 and Table E1). The incidence of VCD was 33%, 23%, and 43% in neonates, infants, and children respectively. The median age of patients with VCD was 21 days (interquartile range [IQR], 11-243 days).

Analysis of Risk Factors for VCD

Twenty-three (24%) patients underwent an aortic arch repair (19 neonates [83%], 3 infants [13%], 1 child [4%]). Of these, VCD occurred in 13 patients (57%) (10

TABLE 2. Intraoperative details and primary surgical procedures*

Variable	Result
Intraoperative	
Transoesophageal echocardiogram utilized	52 (55)
Median sternotomy	69 (73)
Thoracotomy	26 (27)
Use of CPB	56 (59)
Cardioplegia arrest	54 (57)
Antegrade selective cerebral perfusion	24 (25)
Deep hypothermic state <26°	21 (22)
Primary surgical procedures	
Procedure group 1	
PDA ligation/division—primary and associated with other procedures (n = 43 [45%])	
Miscellaneous† + PDA ligation	10
ASO procedure (PDA division)	11
VSD + PDA ligation	9
Isolated PDA ligation	5
RMBTS + PDA ligation	4
PA banding + PDA ligation	4
Procedure group 2	
Coarctation repair without cardiopulmonary bypass (n = 18 [19%])	
Isolated coarctation repair via thoracotomy	17
Coarctation repair associated with right PA reimplantation via sternotomy‡	1
Procedure group 3	
Hypoplastic aortic arch repair via median sternotomy using cardiopulmonary bypass§—primary and associated with other procedure (n = 23 [24%])	
Isolated hypoplastic aortic arch repair	13
Hypoplastic aortic arch repair + ASO	4
Norwood procedure	4
Truncus + IAA repair	1
Interrupted aortic arch repair	1
Procedure group 4	
Vascular ring repair n = 11 (12%) (partial sternal split n = 7, 64%); thoracotomy n = 3, 27%; sternotomy (n = 1 [9%])	
Vascular ring repair	11

Values are presented as n (%). CPB, Cardiopulmonary bypass; PDA, patent ductus arteriosus; ASO, arterial switch operation; VSD, ventricular septal defect; RMBTS, right-modified Blalock-Taussig shunt; PA, pulmonary artery; IAA, interrupted aortic arch repair. *Table 2 demonstrates intraoperative details and primary surgical procedures stratified into 4 groups. †Miscellaneous procedures include biventricular repair of double outlet right ventricle, complete atrioventricular septal defect, partial atrioventricular septal defect, pulmonary atresia with ventricular septal defect, and total anomalous pulmonary venous connection. ‡This patient underwent end–end resection and repair of the coarctation. The procedure was performed via a sternotomy because of concomitant repair of a disconnected right PA; the procedure was performed without the use of CPB. §Hypoplastic aortic arch repair as an isolated or associated procedure was performed in a standardized fashion in our institute utilizing median sternotomy, CPB, and augmentation with a pulmonary homograft patch.

neonates, 2 infants, and 1 child). Pairwise comparison of procedure groups demonstrated that patients undergoing repair of hypoplastic aortic arch were at increased risk of VCD ($P = .002$) (Table E1).

Outcomes

There was no significant difference in the duration of intubation (26 vs 28 hours; $P = .936$), PICU stay (4 vs 4.5 days; $P = .952$) and hospital stay (16 vs 16.5 days; $P = .936$) between those with and without VCD (Table 4).

Feeding

Preoperatively, 37 (39%) patients were nil by mouth, 11 (12%) were predominantly NGT fed (>50% NGT feeds),

11 (12%) were NGT supported (<50% NGT feeds), and 38 (40%) were fully oral fed. Preoperative mode of feeding was similar for those with or without VCD ($P = .303$). Postoperatively, time to first oral trial and liberalization to a patient-specific feeding plan in children with VCD was similar to those without VCD (5 vs 7 days; $P = .91$). A significantly higher percentage of children with vocal cord dysfunction at discharge required supplemental NGT feedings compared with those without VCD (18 out of 30 patients [60%] vs 23 out of 65 patients [36%]; $P = .044$). All children regardless of VCD achieved some degree of oral feeding by the time of discharge and no child was discharged nil by mouth. We did not insert a gastrostomy or perform a tracheostomy in any patient.

TABLE 3. Incidence of vocal cord dysfunction (VCD) stratified by procedure group

Procedure group	VCD–	VCD+	<i>P</i> value*
Procedure group 1: Patent ductus arteriosus ligation/division, primary and associated with other procedures†	35 (81)	8 (19)	.015
Procedure group 2: Coarctation repair via thoracotomy‡	13 (72)	5 (28)	.785
Procedure group 3: Hypoplastic aortic arch repair via sternotomy,§ primary and associated with other procedure	10 (43)	13 (57)	.005
Procedure group 4: Vascular rings	7 (64)	4 (36)	.734

Values are presented as n (%). *P* values in bold indicates statistically significant. *The *P* values were calculated from 4 separate cross-classification tables comparing VCD (yes/no) by each procedure group (yes/no), with the Fisher exact test used to test the null hypothesis of independence. †Procedures include biventricular repair of double outlet right ventricle, complete atrioventricular septal defect, partial atrioventricular septal defect, pulmonary atresia with ventricular septal defect, total anomalous pulmonary venous connection, arterial switch operation, ventricular septal defect closure, pulmonary artery reconstruction, right modified Blalock-Taussig shunt, and interrupted aortic arch repair. ‡This patient underwent end–end resection and repair of the coarctation. The procedure was performed via a sternotomy because of concomitant repair of a disconnected right pulmonary artery; the procedure was performed without the use of cardiopulmonary bypass. §Hypoplastic aortic arch repair as an isolated or associated procedure was performed in a standardized fashion in our institute utilizing median sternotomy, cardiopulmonary bypass, and augmentation with a pulmonary homograft patch.

Follow-up of Patients With VCD (n = 30)

Eight (27%) patients with VCD did not have a follow-up evaluation (6 patients were unable to attend because of COVID-19 related travel restrictions, 1 patient died of an unrelated cause, and 1 patient was awaiting initial follow-up assessment at the time the study concluded).

Twenty-two (73%) patients have had a follow-up investigation. Twenty-one (70%) patients had an LUS (median, 89 days postdischarge) and additionally 14 (47%) underwent FDL (median, 218 days postdischarge). VCD was considered resolved when normal movement of the vocal cords was noted.

Fifteen (68%) patients demonstrated complete resolution of VCD on either LUS or FDL at a median of 97 days postoperatively (IQR, 50-170 days). Seven (32%) had evidence of persistent VCD at a median follow-up of 165 days postoperatively (IQR, 47-200 days) (Figure 3). Six were

neonates; 5 underwent repair of hypoplastic aortic arch. Four children with VCD presented with a respiratory tract infection postdischarge. Three patients required readmission. Two patients were diagnosed with upper respiratory tract infection with respiratory syncytial virus unrelated to aspiration; 1 child presented with right upper lobe collapse that was not believed to be related to aspiration; 1 patient was diagnosed with concomitant aspiration on admission. This patient underwent a video fluoroscopic swallow study that demonstrated reflux and disorganized swallow but no aspiration. Following recovery, the child returned to full oral feeds. There were no other readmissions to hospital for any child with VCD.

Diagnostics (n = 60)

Sixty patients underwent trimodal evaluation (ie, assessment by a speech pathologist, LUS, and FDL). Thirty-four (36%) patients did not undergo FDL (COVID-19, n = 22;

TABLE 4. Hospital stay and feeding outcomes

Outcomes	VCD (n = 30)	No VCD (n = 65)	<i>P</i> value
Intubation (h)	28 (8.5-77)	26 (7-102)	.936
Hospital stay (d)	16.5 (9.5-26.8)	16 (7-26)	.703
PICU stay (d)	4.5 (2-7)	4 (1-8)	.952
Feeding			
Time to first oral feed (d)	7 (2.3-9.5)	5 (3-10)	.91
Requirement for NGT feeding	24 (80)	56 (86)	.547
Supplementary NGT feeding at discharge	18 (60)	23 (36)	.044
Days of NGT feeding (d)	32.5 (14.3-14.8)	19 (7-38)	.25
Mode of feeding at discharge			
Full oral feeds	12 (40)	42 (65)	.043
NGT supported: >50% oral feeds	11 (37)	19 (29)	.64
NGT predominant: <50% oral feeds	7 (23)	4 (6)	.03

Values are presented as median (interquartile range) or n (%). *P* values in bold indicates statistically significant. VCD, Vocal cord dysfunction; PICU, pediatric intensive care unit; NGT, nasogastric tube.

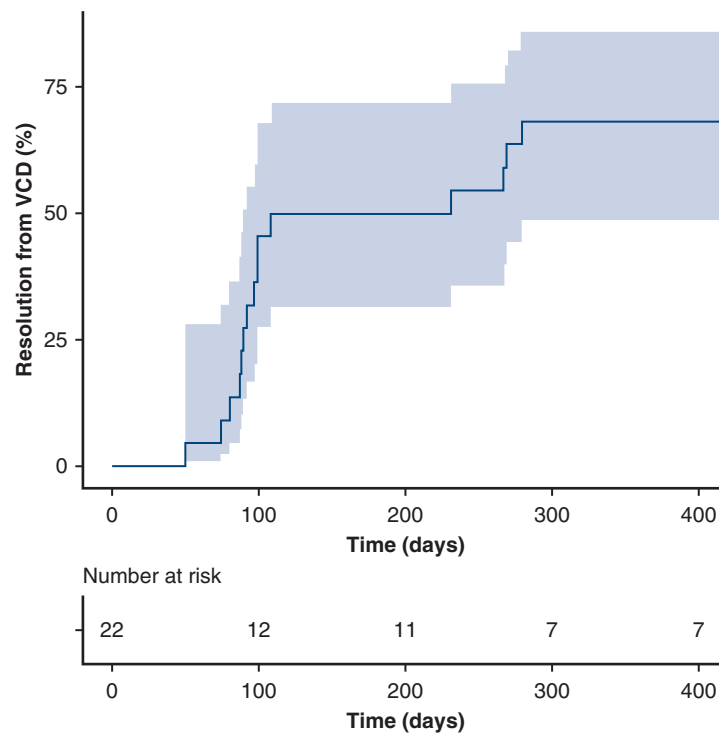


FIGURE 3. Kaplan-Meier curve demonstrating resolution of vocal cord dysfunction (VCD) on either laryngeal ultrasound (LUS) or flexible direct laryngoscopy (FDL). As per the study protocol, patients undergo LUS at 3 months, followed by FDL at 6 months. Patients were censored at the earliest investigation demonstrating return of normal movement of the vocal cords. Eight (27%) patients with VCD did not have a follow-up evaluation (6 patients were unable to attend because of COVID-19–related travel restrictions, 1 patient died of an unrelated cause, and 1 patient was awaiting initial follow-up assessment at the time the study concluded). Of the 73% (22 out of 30) of patients with VCD who underwent a follow-up evaluation during the study period, complete resolution of VCD occurred in two-thirds of patients at a median of 3 months. VCD, Vocal cord dysfunction.

parents declined FDL, $n = 5$; patient discharged before ENT evaluation, $n = 4$; and patient noncompliant during the procedure, $n = 3$). One patient did not undergo LUS (radiographer unavailability before discharge). No parent or child declined LUS.

The sensitivity, specificity, positive and negative predictive values, and Cohen kappa coefficient of speech pathology evaluation in conjunction with LUS compared with FDL for VCD were 91%, 84%, 78%, 94%, and 0.73, respectively (Tables E2 and E3).

Assessment by a speech pathologist alone or LUS alone demonstrated lower sensitivity and negative predictive value compared with combined modalities. Differences were not statistically significant (Table E1). There were 2 false negatives and 6 false positives for combined speech pathology assessment and LUS that were not statistically associated with any patient or surgical factors.

Silent VCD

Four (13%) patients with VCD were asymptomatic when assessed by the speech pathologist. Of these, 2 patients were also incorrectly diagnosed without VCD on LUS. One of these patients had complete resolution of VCD on follow

up LUS and FDL, 1 patient was lost to follow-up, 1 patient is awaiting follow-up investigation and is fully orally fed, and 1 patient had persistent VCD at 226 days postoperatively. This patient remains asymptomatic with no voice abnormality and is fully orally fed.

DISCUSSION

Incidence

Our study demonstrated that one-third of all children undergoing surgery for congenital heart disease with increased risk of injury to the RLN developed VCD postoperatively. The reported incidence of VCD ranges from 4.5% to 59%, reflecting the variability in study cohorts, screening methods, and data collection.^{1,2} One report with inclusion criteria like ours reported a lower incidence of 12.1%. However, this was a retrospective study with screening of only symptomatic patients.¹⁸ The overall incidence of VCD in neonates in our cohort was 33%, which is higher than the reported 6.9% from a cross-sectional database analysis of all neonates undergoing surgery for CHD in the United States.⁷ The incidence of VCD ranged from 18% to 56% and varied according to procedure group with the highest incidence in patients undergoing aortic

arch repair via median sternotomy. However, this retrospective study did not involve standardized screening, and the incidence is likely to be under reported and prone to institutional bias.

Risk Factors

Studies have identified prematurity, younger age, lower weight, utilization of circulatory arrest, monopolar diathermy, preoperative intubation, cuffed endotracheal intubation, transoesophageal echocardiography, and comorbidities as risk factors for developing VCD.^{2,6,18,19} Our data demonstrated that repair of hypoplastic aortic arch as an isolated or combined procedure carried a significantly higher risk of VCD. VCD occurred in 56% of patients undergoing repair of the aortic arch in our study. This is similar to previous reports (48%-59%).^{2,3} Arch repair in our institute is performed in a standardized fashion utilizing cardiopulmonary bypass, mobilization of the arch and descending thoracic aorta, and augmentation with a pulmonary homograft patch. This involves unavoidable mobilization of the RLN and potential stretching of the nerve by the homograft patch. The risk factors identified on univariable analysis in our study, antegrade cerebral perfusion and deep hypothermia, are both associated with our technique of aortic arch repair.

Natural History

Two-thirds of our patients with VCD demonstrated resolution on imaging at a median of 3 months. Eleven of the 15 patients recovered by 3 months, and the remaining 4 recovered by 7 months. This is similar to a previous report.²⁰ One-third of our cohort remain with unresolved VCD. The long-term outcome of patients with persistent VCD remains unclear. Mery and colleagues²¹ reported 97% resolution at 6-year follow-up. Notably, theirs was a retrospective cohort and the diagnosis of resolution was based on clinical examination and not on an imaging modality. Six of the 7 patients with persistent VCD in our study were receiving full oral feeds 1 year postoperatively suggesting that compensation by the unaffected vocal cord is effective in preventing aspiration.

Consequences of VCD

A national population-based study exploring the resource impact of RLN injury in congenital heart surgery reported a significantly longer hospitalization (34 vs 18 days) and a mean adjusted cost increase of US\$34,123 per patient admission in those with VCD.⁷ In our study, children with VCD did not have a prolonged duration of intubation, PICU stay, or hospital stay, compared with those without VCD. This may be explained by our policy to attempt oral feeding in neonates only after discharge from the PICU. The comparable duration of hospital stay may reflect our experience with a structured approach to assessment

and feeding on the ward by the speech pathology team even before the commencement of the study.

Feeding

Feeding in children undergoing cardiac surgery is not only affected by the function of the vocal cords but also by other complex sensory and motor mechanisms of laryngopharyngeal function.²² Barriers to oral feeding are frequently present preoperatively, and include prostaglandin E1 dependence, risk of necrotizing enterocolitis, cardiac failure, and trachea-oesophageal compression by vascular rings.¹ Although VCD remains a major contributor to feeding challenges with only 21% to 50% achieving oral feeds on discharge in other studies,^{4,23-26} it is not the whole story. In previous studies, more than half of neonates with feeding difficulties had no VCD on direct laryngoscopy, whereas a quarter of those with VCD had no swallowing dysfunction.^{22,27} A recent systematic review reported an inability to maintain oral diet in 14% to 100% of children with VCD, and 11% to 61% of patients without VCD following cardiac surgery.²⁰ The multifactorial nature of the barriers to establishing full oral feeding were evident in our cohort, with 44% of children without VCD requiring NGT supplementation of feeding at hospital discharge.

We found that significantly more children with VCD required NGT feeding at discharge, but that all these children were able to achieve some degree of oral feeding. Forty percent were able to achieve full oral feeding before discharge, and all but 1 patient with VCD were able to ultimately achieve full oral feeding safely in a similar timeframe to those without VCD. This can be attributed to a protocol driven care with immediate implementation of a patient-specific feeding plan. Daily tailoring of the feeding plan to the individual child may have further mitigated aspiration risk and allowed progression. However, we based our evaluation on clinical examination and did not systematically evaluate patients for aspiration with a modified barium swallow. Consequently, it is possible that aspiration was missed in some patients and consideration should be given to performing a modified barium swallow especially in high-risk patients based on institutional preference.

Although others report readmission due to feeding difficulties and poor weight gain up to 7 times more common in children with VCD, we did not find our patients to require readmission due to feeding difficulties.²³ This may reflect ongoing evaluation by the speech pathology team with modification of the feeding plan as necessary after discharge.

The high risk of feeding difficulties and aspiration and the related attrition in hypoplastic left heart syndrome patients with VCD has led some units to perform preemptive gastrostomy.²⁵ Although this has been shown to improve survival to the second stage, it exposes patients to additional risks, including peritonitis, tube dislodgement, and the need for further intervention.²⁵ Our study was not powered to analyze

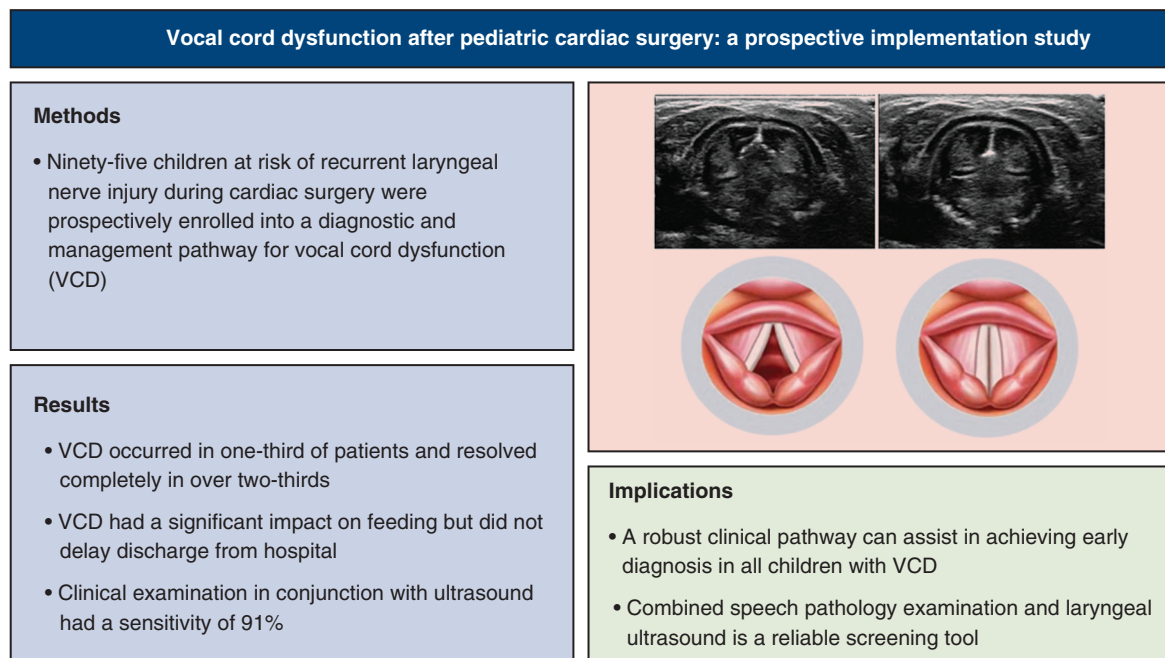


FIGURE 4. The key features and findings of a prospective implementation study regarding vocal cord dysfunction (VCD) in children after cardiac surgery.

univentricular patients because 90% of our cohort underwent biventricular repair. As an institutional preference, we prefer a trial of NG feeding with outpatient monitoring for all diagnoses before considering a gastrostomy. Furthermore, all patients undergoing a Norwood procedure are discharged into a home monitoring program involving accommodation close to the hospital. Parents undergo structured training and patients requiring NG feeding are closely monitored with frequent speech pathology assessment.

Diagnosis of VCD

In children, laryngeal cartilages are not yet calcified permitting clearer identification of the endolaryngeal structures.²⁸ The good consistency demonstrated between LUS and FDL in both normal and pathological vocal cords has prompted the adoption of LUS as a primary modality for routine postoperative evaluation.^{2,11} Additionally, LUS has been shown to produce less hemodynamic compromise, an important consideration in neonates following complex cardiac procedures.^{13,29} LUS also has higher parental acceptability and comfort. In our study, no child or parent declined LUS in comparison to 5 patients whose parents declined FDL and 4 children who were noncompliant with FDL.

Our results corroborate the value of LUS as a diagnostic modality but highlight the importance of combined assessment by a speech pathologist. We found a sensitivity of 91% and a specificity of 84% when both were used in conjunction. The sensitivity of LUS alone was 74%, and, in our unit, LUS is not used in isolation to identify

postoperative VCD. A previous report demonstrated a sensitivity of 95% with LUS in a homogenous cohort of patients undergoing repair of aortic arch.² In contrast, our patient group was heterogenous regarding age, diagnosis, and procedure. FDL is performed only when the findings of the LUS are equivocal and there is a strong suspicion of VCD based on speech pathologist assessment.

Silent VCD

Thirteen percent of children with VCD in our cohort had no clinical signs. Previous studies have reported a 25% to 50% incidence of VCD with normal swallow feeding assessments.^{21,22} All patients with silent VCD in our cohort achieved full oral feeding. Identification of these patients remains important, given the reported increased risk of aspiration in the presence of VCD, especially in neonates and infants.³

Limitations

The COVID-19 pandemic limited access to FDL and ENT follow-up for a 3-month period. Consequently, the sample of patients who underwent trimodal assessment was smaller than anticipated (reduced from 95 to 60). The loss in statistical power decreased the precision with which we could measure the sensitivity and specificity (reflected in the wide confidence limits). A small number of patients who did not undergo FDL were included in the risk factor analysis, possibly leading to minor misclassification in VCD status. There were only 4 patients who underwent a Norwood procedure in our study cohort. Norwood patients

are known to be at higher risk of aspiration and the small number of patients in our study could have influenced the requirement for gastrostomy tube placement. However, we do not treat Norwood patients differently. They are all assessed and managed using the same structured pathway as patients with a biventricular circulation. Lastly, 6 patients diagnosed with VCD after the initial surgery were unable to attend follow-up due to national travel restrictions and parental reluctance to travel during the pandemic.

CONCLUSIONS

Our key conclusions are demonstrated in **Figure 4**. Overall, VCD occurred in one-third of patients undergoing cardiothoracic surgery involving intervention on the aortic arch, ductus arteriosus, ligamentum arteriosum, and vascular rings. The incidence of VCD was different in each procedure group and ranged from 18% to 56%. More than half of patients who underwent surgery on the aortic arch developed VCD. Of the 73% of patients with VCD who underwent a follow-up evaluation during the study period, complete resolution of VCD occurred in two-thirds of patients at a median of 3 months. VCD did not delay discharge from the PICU or hospital. VCD adversely influenced feeding during the postoperative period. Full oral feeding was achieved in 40% of patients before discharge. The combination of assessment by a specialist speech pathologist and LUS is more patient friendly and can be used to diagnose VCD accurately in most cases.

Webcast

You can watch a Webcast of this AATS meeting presentation by going to: https://aats.blob.core.windows.net/media/21%20AM/AM21_C04/AM21_C04_04.mp4.



Conflict of Interest Statement

The authors reported no conflicts of interest.

The *Journal* policy requires editors and reviewers to disclose conflicts of interest and to decline handling or reviewing manuscripts for which they may have a conflict of interest. The editors and reviewers of this article have no conflicts of interest.

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Key Words: vocal cord dysfunction, pediatric cardiac surgery, recurrent laryngeal nerve

APPENDIX E1. STATISTICAL METHODOLOGY

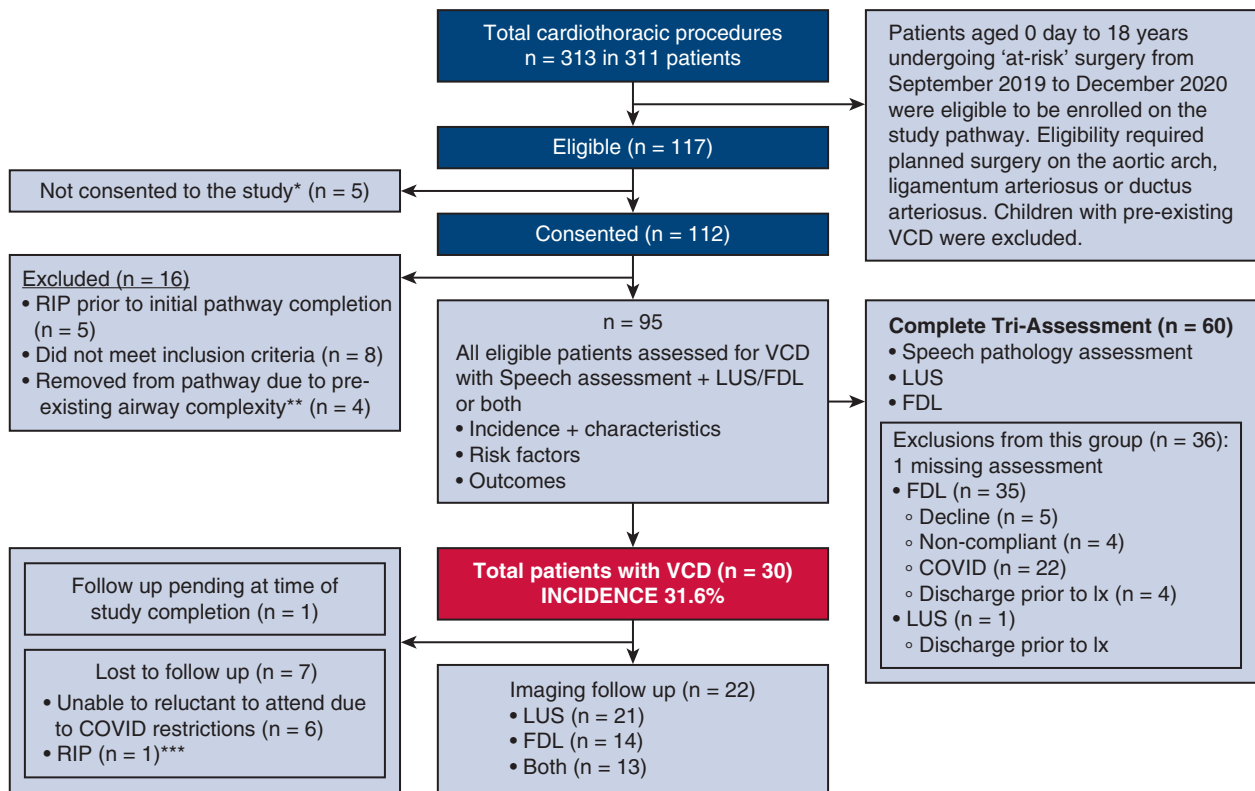
Initially, a global test was performed (Fisher exact) to test the null hypothesis that vocal cord palsy (VCP) status was independent of procedure category. Because the null hypothesis was rejected ($P = .017$), we next compared each procedure group against each other by VCP status (Fisher test). These pairwise comparisons found patients

in procedure group 3 were at a statistically and clinically significant increased risk of VCP compared with procedure 1 ($P = .002$). Our data also indicated those in procedure group 3 were at increased risk compared with those in procedure groups 2 and 4, but due to the small numbers in these latter groups, the null hypothesis was not rejected.

TABLE E1. Statistical analysis of procedure groups

Procedure group (n = 95)	VCP-	VCP+	Pairwise comparison <i>P</i> value*			
			Procedure 1	Procedure 2	Procedure 3	Procedure 4
Procedure group 1: PDA ligation/division – primary and associated with other procedure† (n = 43 [45%])	35 (53.9)	8 (26.7)	Procedure 1	.499	.002	.237
Procedure group 2 (coarctation repair via thoracotomy‡), n = 18 (19%)	13 (20)	5 (16.7)	Procedure 2		.113	.694
Procedure group 3: Hypoplastic aortic arch - primary and associated with other procedure§ (n = 23 [24%])	10 (15.4)	13 (43.3)	Procedure 3			.464
Procedure group 4: Vascular rings (n = 11 [12%])	7 (10.8)	4 (13.3)	Procedure 4			
Global test*	.017					

Values are presented as n (%). *P* value in bold indicates statistically significant. *VCP*, Vocal cord palsy. *Fisher exact test. †Procedures include biventricular repair of double outlet right ventricle, complete atrioventricular septal defect, partial atrioventricular septal defect, pulmonary atresia with ventricular septal defect, total anomalous pulmonary venous connection, arterial switch operation, ventricular septal defect closure, pulmonary artery reconstruction, right modified Blalock-Taussig shunt, and interrupted aortic arch repair. ‡This patient underwent concomitant repair of right pulmonary artery disconnection; the procedure was performed via median sternotomy. §Hypoplastic aortic arch repair as an isolated or associated procedure was performed in a standardized fashion in our institute utilizing median sternotomy, cardiopulmonary bypass, and augmentation with a pulmonary homograft patch (n = 21 out of 23).



* Five patients were not consented for the study: a) One neonate underwent emergency repair of an obstructed TAPVC with PDA ligation. The patient could not be weaned from cardiopulmonary bypass and was placed onto ECMO support in the operating theatre. The baby died without being weaned from ECMO; b) One child had severe autism and significant healthcare related anxiety in whom we decided to limit the number of postoperative investigations to facilitate clinical care; c) One neonate with complex social circumstances and language barriers where additional informed consent for the study was deemed to be challenging and burdensome for the family; d) Two neonates who underwent emergency repair of obstructed TAPVC and PDA ligation were inadvertently not consented due to oversight by the study team.

** Patients removed from pathway due to pre-existing conditions: multilevel airway complexities secondary to a) Undiagnosed genetic phenotype with pre-existing laryngomalacia, microcephaly, cleft lip and palate, micrognathia, macroglossia, anterior anus, hypertelorism; b) Pierre-Robin + 22q11 deficiency; c) Cri Du chat syndrome; d) Xq24 deletion UBE2A deficiency with significant pharyngomalacia and upper airway obstruction.

*** death unrelated to VCP

FIGURE E1. Consolidated Standards of Reporting Trials flow diagram.

TABLE E2. Sensitivity, specificity, positive predictive value, negative predictive value, and Cohen kappa coefficient for combined speech pathology and laryngeal ultrasound (LUS) assessment, LUS alone isolation, and speech pathology (SP) assessment alone

Variable	Combined LUS + SP	LUS alone	SP examination alone
Sensitivity	0.91 (0.72-0.99)	0.74 (0.52-0.90)	0.83 (0.61-0.95)
Specificity	0.84 (0.68-0.94)	0.92 (0.78-0.98)	0.92 (0.79-0.98)
Positive predictive value	0.78 (0.58-0.91)	0.85 (0.62-0.97)	0.86 (0.65-0.97)
Negative predictive value	0.94 (0.80-0.99)	0.85 (0.70-0.94)	0.90 (0.76-0.97)
Cohen kappa	0.73 (0.48-0.98)	0.67 (0.42-0.92)	0.75 (0.50-1.00)

Values are presented as value (95% CI).

TABLE E3. Distribution of test positive/negative on the three diagnostic tests v the actual outcome according to flexible direct laryngoscopy (FDL)

Test	Outcome: FDL	
	Yes	No
Combined LUS + SP		
Positive	21	6
Negative	2	31
LUS		
Positive	17	3
Negative	6	34
SP		
Positive	19	3
Negative	4	35

LUS, Laryngeal ultrasound; SP, speech pathology.